CASE REPORT

Dural arteriovenous fistula presenting with acute subdural haematoma showing impending cerebral herniation

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SUMMARY

A dural arteriovenous fistula (DAVF) presenting with acute subdural haematoma (ASDH), which were not related to head injury, is rare. A 61-year-old woman was transported by ambulance because of deterioration of consciousness. On admission, she was comatose with anisocoria. Emergent CT demonstrated a severe midline shift associated with a left ASDH and an additional left occipital intracerebral haematoma, both of which had no continuity with each other. MRI showed flow void signs in the left occipital lobe. Because of the impending cerebral herniation, an emergent evacuation of the ASDH and external decompression was performed. Subsequent evaluation revealed a DAVF at the left occipital convexity near the confluence with retrograde leptomeningeal venous reflux and venous ectasia (Cognard type III DAVF). The patient underwent endovascular treatment for the DAVF involving transarterial embolisation using coils and *N*-butyl cyanoacrylate with complete obliteration. Her further clinical course was uneventful and discharged after cranioplasty.

BACKGROUND

A dural arteriovenous fistula (DAVF) presenting with an acute subdural haematoma (ASDH) is rare. We report the case of a DAVF that presented with an intracerebral haematoma (ICH) and an ASDH, which were not related to head injury. Only 19 similar cases have been reported, including our case (table 1).^{1–15}

Because of its sparsity, a standard treatment strategy has not been established yet, and treatment is selected according to the condition of each patient. In this report, possible mechanisms for the pathogenesis of ASDH associated with DAVF and the treatment strategy are discussed on the basis of our case and previous reports.

CASE PRESENTATION

A 61-year-old woman presented with acute onset headache followed by unconsciousness and was admitted to our hospital. There was no history of head injury or other causative diseases. On admission, she was comatose (Glasgow coma scale score of 6) and showed anisocoria (R<L). Physiological evaluation showed a high blood pressure of 190/110 mm Hg and decreased oxygen saturation of 92% because of mandibular breathing. CT revealed an ICH in the left occipital lobe combined with a

large ASDH in the left cerebral hemisphere with a marked midline shift (figure 1A). Emergent MRI revealed flow void signs close to the ICH, which suggested a DAVF. The two haematomas had no continuity (figure 1B). Because the patient's condition indicated cerebral herniation, she underwent emergent craniotomy for the removal of ASDH and external decompression. As we thought that it was not possible to manage ASDH and possible DAVF simultaneously, craniotomy was not extended to occipital region and the ICH was untreated, while reddish cortical veins were observed in the operative field at the parieto-occipital cortical surface.

Her consciousness immediately recovered after the surgery; however, postoperative CT showed a slight increase in the size of ICH (figure 1C). Therefore, we immediately resumed general anaesthesia with propofol for strict blood pressure control to prevent re-bleeding even though the intracranial pressure was not so high by observation of the bone window. Digital subtraction angiography (DSA), performed on day 4 after the surgery, revealed a DAVF at the left occipital cranial vault near the confluence, which was fed by the left posterior meningeal artery originating from the left vertebral artery (figure 2A, B) and the neuromeningeal branch of the left ascending pharyngeal artery (figure 2C, D). It drained into the left internal occipital vein with venous ectasia measuring 2.5 mm in diameter (figure 2E). Based on these findings, the lesion appeared to be a highly dangerous DAVF with retrograde leptomeningeal venous drainage (RLVD) and without sinus drainage. It was classified as Cognard type III,¹⁶ indicating a high risk of intracranial rebleeding, and early curative treatment is considered to be required.¹⁶ She underwent emergent endovascular treatment under general anaesthesia on the day after DSA. Transarterial embolisation (TAE) of the meningeal branch of the vertebral artery was initially performed via the left vertebral artery using platinum detachable coils (figure 3A, B). Then, TAE of the neuromeningeal branch of the ascending pharyngeal artery was performed using 20% *N*-butyl cyanoacrylate diluted with lipiodol (figure 3C, D) and complete obliteration of the DAVF was achieved.

OUTCOME AND FOLLOW-UP

The postoperative course was uneventful, and cranioplasty was performed 1 month after the initial

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Table 1 Dural arteriovenous fistula presenting with an acute subdural haematoma: previous reports

Case, Author	Age	Sex	Presenting symptoms	Location of DAVF	Location of haematoma	Cognard classification ¹⁶	Treatment	Outcome
Nakagawa <i>et al</i> ¹	60	M	Headache, DOC	Frontal base	ASDH+frontal ICH	IV	Surg	Improved
Baskaya <i>et al</i> ²	51	F	Headache	Frontal base	ASDH	undetermined	Surg	Improved
Duffau <i>et al</i> ³	55	M	Headache, monoparesis	Unknown	SDH	III	End ->Surg	Improved
Duffau <i>et al</i> ³	64	M	Headache, hemiplegia	Temporal	ASDH+temporal ICH	III	End ->Surg	Died
Duffau <i>et al</i> ³	64	M	DOC, hemiparesis	Frontal	ASDH+frontal ICH	IV	Surg	Improved
Duffau <i>et al</i> ³	56	F	Headache	Occipital	ASDH	III	Surg	Died
Sugawara <i>et al</i> ⁴	67	M	DOC, neurological deficits	Frontal base	ASDH+frontal ICH	IV	Surg	Improved
Kagawa <i>et al</i> ⁵	65	F	Headache, DOC	Cavernous sinus	ASDH+SAH	IV	Surg ->End	Died
Dietrich <i>et al</i> ⁶	68	F	DOC, hemiparesis	Occipital	ASDH+ICH	III	End	Improved
Kominato <i>et al</i> ⁷	42	F	Headache	Falx	ASDH	undetermined	None	Died
Tanei <i>et al</i> ⁸	75	M	DOC	Frontal base	ASDH+frontal ICH	undetermined	Surg	Improved
Matsuzaki <i>et al</i> ⁹	66	M	Cognitive dysfunction	Occipital	ASDH+temporo occipital ICH	IV	End	Improved
Kohyama <i>et al</i> ¹⁰	60	M	Headache	Low convexity	ASDH	IIa	End ->Surg	Improved
Kohama <i>et al</i> ¹¹	58	M	Headache	Frontal base	ASDH+SAH	IV	Surg	Improved
Kitazone <i>et al</i> ¹²	68	M	Headache	Occipital	ASDH+occipital ICH	IV	Surg	Improved
Ogawa <i>et al</i> ¹³	27	M	Headache	Convexity (near SSS)	ASDH	III	Surg	Improved
Saito <i>et al</i> ¹⁴	56	M	DOC	Occipital	ASDH+occipital ICH	IV	End ->Surg	Improved
d'Angelo <i>et al</i> ¹⁵	60	F	Headache, DOC	Frontotemporal	ASDH	IV	End	Improved
Present case	61	F	DOC	Occipital	ASDH+occipital ICH	III	Surg ->End	Improved

ASDH, acute subdural haematoma; DAVF, dural arteriovenous fistula; DOC, deterioration of consciousness; End, endovascular treatment; ICH, intracerebral haematoma; SSS, superior sagittal sinus; Surg, surgery.

surgery. She was discharged without any neurological deficits 2 weeks after cranioplasty. She has been neurologically intact and follow-up MRI/MRA shows no recurrence of the DAVF at a 3-year follow-up.

DISCUSSION

The incidence of a DAVF presented with intracranial haemorrhage as the initial symptom has been reported to be 1.5%–2.0%.¹⁷ Most cases have RLVD, often combined with venous ectasia. They are usually classified as Cognard types IIb, III and IV.¹⁶ The annual risk of intracranial haemorrhage has been reported to be >10%.¹⁸ In most patients, an ICH or subarachnoid haemorrhage (SAH)

occurs, and those with an ASDH are extremely rare. To our knowledge, only 19 (men, 12; women, 7) patients, including our patient, have been reported since 1990 (table 1).^{1–15} The mean patient age was 59.1 ± 10.6 years. Additionally, more than half of the patients exhibited disturbance of consciousness, requiring emergent treatment. The common locations of ASDHs were the frontal and occipital regions. Of the 19 patients, 5 were Cognard type III, 10 were type IV, 1 was type IIa and 3 were unknown. Only ASDH occurred in seven patients, whereas ICH or SAH was simultaneously noted in the other patients. The cause of intracranial haemorrhage is believed to be the rupture of the cortical vein of RLVD or venous ectasia, leading to ICH, SAH or both because of venous hypertension.¹⁹ The mechanism of ASDH development has not

Figure 1 (A) CT on admission showing an ASDH in the left cerebral hemisphere with a remarkable midline shift. An ICH is also seen in the left occipital lobe. (B) MRI on admission shows the same haematomas identified on CT. The continuity of both haematomas is not observed. Flow void signs are noted in the left occipital lobe adjacent to the ICH. (C) CT after initial craniotomy showing a complete evacuation of ASDH, an external decompression and an alleviation of the midline shift. A slight increase in the size of ICH is noted. ASDH, acute subdural haematoma; ICH, intracerebral haematoma.

Figure 2 (A, B) Left VAG (A: anteroposterior view, B: lateral view) showing a DAVF at the left occipital cranial vault superiolateral to the confluence, fed by the left posterior meningeal artery from the left vertebral artery, draining into the left internal occipital vein. Right ECAG (C: anteroposterior view, D: lateral view) showing DAVF as noted on left VAG, fed by the meningeal branch. An enlarged draining vein with venous ectasia is noted. (C, D) Three-dimensional digital subtraction angiography of the left external carotid artery (E: lateral view) clearly showing the locations of the DAVF, enlarged draining vein (arrow) and venous ectasia (arrowhead). DAVF, dural arteriovenous fistula; ECAG, external carotid angiography; VAG, vertebral angiography.

Figure 3 (A) Selective injection of the left posterior meningeal artery (anteroposterior view) during endovascular treatment. The left posterior meningeal artery was embolised here with platinum coils. (B) Left vertebral angiography (lateral view) during endovascular treatment showing a disappearance of the DAVF. (C) Selective injection of the neuromeningeal branch of the right ascending pharyngeal artery (lateral view) during endovascular treatment. The artery was embolised from this point using *N*-butyl cyanoacrylate with sufficient penetration into the lesion. (D) Right external carotid angiography (lateral view) showing a disappearance of the DAVF. DAVF, dural arteriovenous fistula.

been well documented, except a report mentioning that the subpial cortical vein with laceration of the arachnoid causes ASDH.¹⁴ In our patient, both ICH and ASDH were observed, but there was no continuity between them. We assumed that the ICH initially occurred in the medial occipital lobe and was caused by a rupture of venous ectasia. Subsequently, the enhancement of venous hypertension induced the secondary rupture of the cortical vein at the junction of the dural sinus, leading to ASDH.

Among the 19 reported patients of DAVF with ASDH, prognosis was favourable in 14, death occurred in 4 and the prognosis was unclear in 1. Because the degree of ASDH is a crucial factor for prognosis, the treatment strategy depends on its severity. If intracranial hypertension is not manageable with conservative therapy, then an emergent evacuation of the ASDH is mandatory. In contrast, an early curative treatment of DAVF is desired because it has been reported that DAVF with intracranial haemorrhage can easily rebleed and that 35% of the patients show rebleeding within 2 weeks of the onset.³

Among the 19 reported patients, 18 underwent surgical treatment. Craniotomy for treating both ASDH and DAVF was performed in nine patients at a single stage. Three patients underwent endovascular treatment for DAVF and received medication only for ASDH. In six patients, craniotomy for ASDH and endovascular treatment for DAVF were performed in combination. Among these patients, four underwent endovascular treatment initially followed by craniotomy, whereas the remaining two patients underwent craniotomy initially followed by endovascular treatment. However, craniotomy failed in one patient due to difficulty in haemostasis, and the patient was lost without endovascular treatment.⁵

Bleeding during the evacuation of ASDH may become uncontrollable if DAVF is not obliterated. Simultaneous management by direct surgery may be desirable if both evacuation of ASDH and curative procedure for DAVF, involving the coagulation of RLVD outflow, can be performed in the same operative field.⁴ However, simultaneous treatment may not be feasible if the surgical field of haematoma evacuation is distant from that of DAVF treatment, as in our case. In such a situation, a two-stage treatment approach is inevitable, with the risk of uncontrollable bleeding. If the ASDH can be medically treated or evacuation can be delayed, then the curative treatment of DAVF through either direct surgery or endovascular treatment can be initially performed. If the ASDH drastically worsens and an emergent decompression is required,

then it is inevitable to postpone the curative treatment for DAVF before craniotomy. A hybrid operating room may help decrease the interval between craniotomy for ASDH and DAVF treatment.

Learning points

- ▶ Intracranial dural arteriovenous fistula (DAVF) may rarely present with acute subdural haematoma (ASDH).
- ▶ A case of DAVF presented with ASDH may require an emergent treatment for impending cerebral herniation with curative treatment for DAVF.
- ▶ The selection of surgical treatments and their order for ASDH and DAVF require careful consideration according to the location of both lesions and the condition of the patient.

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REFERENCES

- 1 Nakagawa K, Takasato Y, Ito Y. [A case of dural arteriovenous malformation in the anterior cranial fossa presenting with intracranial hemorrhage]. *No To Shinkei* 1994;46:387–91.
- 2 Başkaya MK, Suzuki Y, Seki Y, Negoro M, et al. Dural arteriovenous malformations in the anterior cranial fossa. *Acta Neurochir* 1994;129:146–51.
- 3 Duffau H, Lopes M, Janosevic V, et al. Early rebleeding from intracranial dural arteriovenous fistulas: report of 20 cases and review of the literature. *J Neurosurg* 1999;90:78–84.
- 4 Sugawara T, Seki H, Wada T, et al. Impending herniation due to intracerebral and subdural hematoma with dural arteriovenous fistula in the anterior fossa. *Nihon Kyokyu Igakukai Zasshi* 2000;11:219–24.
- 5 Kagawa K, Nishimura S, Seki K. [Cavernous sinus dural arteriovenous shunt presenting with subarachnoid hemorrhage and acute subdural hematoma: a case report]. *No Shinkei Geka* 2001;29:457–63.
- 6 Dietrich U, Wanke I, Asgari S, et al. [Dural arteriovenous fistulas with intracranial hemorrhage: diagnostic and therapeutic aspects]. *Zentralbl Neurochir* 2003;64:12–18.
- 7 Kominato Y, Matsui K, Hata Y, et al. Acute subdural hematoma due to arteriovenous malformation primarily in dura mater: a case report. *Leg Med* 2004;6:256–60.
- 8 Tanei T, Fukui K, Wakabayashi K, et al. Dural arteriovenous fistula in the anterior cranial fossa: four case reports. *Neurol Med Chir* 2008;48:560–3.
- 9 Matsuzaki T, Shibuya T, Suma T, et al. [A case of dural arteriovenous fistula mimicking isolated transverse-sigmoid sinus successfully treated via the transvenous route]. *No Shinkei Geka* 2009;37:1221–5.
- 10 Kohyama S, Ishihara S, Yamane F, et al. Dural arteriovenous fistula presenting as an acute subdural hemorrhage that subsequently progressed to a chronic subdural hemorrhage: case report. *Minim Invasive Neurosurg* 2009;52:36–8.
- 11 Kohama M, Nishimura S, Mino M, et al. Anterior cranial fossa dural arteriovenous fistula with bilateral cortical drainers—case report. *Neurol Med Chir* 2010;50:217–20.
- 12 Kitazono M, Yamane K, Toyota A, et al. [A case of dural arteriovenous fistula associated with subcortical and subdural hemorrhage]. *No Shinkei Geka* 2010;38:757–62.
- 13 Ogawa K, Oishi M, Mizutani T, et al. Dural arteriovenous fistula on the convexity presenting with pure acute subdural hematoma. *Acta Neurol Belg* 2010;110:190–2.
- 14 Saito A, Kawaguchi T, Sasaki T, et al. A case of dural arteriovenous fistula presenting as acute subdural hematoma. *Case Rep Neurol* 2014;6:122–5.
- 15 d'Angelo VA, Monte V, Scialfa G, et al. Intracerebral venous hemorrhage in "high-risk" carotid-cavernous fistula. *Surg Neurol* 1988;30:387–90.
- 16 Cognard C, Gobin YP, Pierot L, et al. Cerebral dural arteriovenous fistulas: clinical and angiographic correlation with a revised classification of venous drainage. *Radiology* 1995;194:671–80.
- 17 Maiuri F, Iaconetta G, Sardo L, et al. Dural arteriovenous malformation associated with recurrent subdural haematoma and intracranial hypertension. *Br J Neurosurg* 2001;15:273–6.

18 van Rooij WJ, Sluzewski M, Beute GN. Dural arteriovenous fistulas with cortical venous drainage: incidence, clinical presentation, and treatment. *AJNR Am J Neuroradiol* 2007;28:651–5.

19 Okamoto S, Harada J, Nishijima M, *et al.* Dural arteriovenous fistula in the anterior cranial fossa associated with intracerebral hemorrhage in the temporal lobe : a case report. *Japanese Journal of Neurosurgery* 2001;10:807–11.

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